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Primary retroperitoneal hydatid disease mimicking retroperitoneal malignant tumor

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Summary
Background: Hydatid disease (HD) is endemic in many parts of the world. It may develop in almost any part of the body; the liver is the organ most frequently involved. HD in an unusual location may make differential diagnosis difficult. Isolated retroperitoneal HD is extremely rare.
Case report: We report herein a case of retroperitoneal HD mimicking retroperitoneal malignant tumor.
Conclusions: HD should be considered in the differential diagnosis of all cystic masses in all anatomic locations, especially in regions of the world where the disease is endemic.

Introduction

Hydatid disease (HD), known since the time of Hippocrates, still constitutes a serious public health problem in areas in which it is endemic. It is caused by the parasite *Echinococcus granulosus* and is usually found in the liver and lungs, but can also develop anywhere in the body including spleen, kidney, pancreas, peritoneum, retroperitoneum, central nervous system, soft tissues, and the breast.[1—3]

There are no specific local or general symptoms and signs of HD, and it is often diagnosed following incidental findings on radiographic examination for unrelated complaints.[2—4] Imaging modalities such as ultrasound (US), computerized tomography (CT), and magnetic resonance imaging (MRI) may reveal a calcified cyst wall and microcalcifications within daughter cysts and varying fluid densities between cysts and surrounding organs.[2,4] Despite characteristic imaging findings, HD in unusual anatomic locations may make differential diagnosis difficult. The combination of clinical history, imaging findings, and serologic test results usually aids in the diagnosis.[1,2]

Isolated retroperitoneal HD is extremely rare and is usually secondary to the involvement of other organs (especially the liver) or to surgery. We report herein a case of hydatid disease of the retroperitoneum without any other organ involvement.

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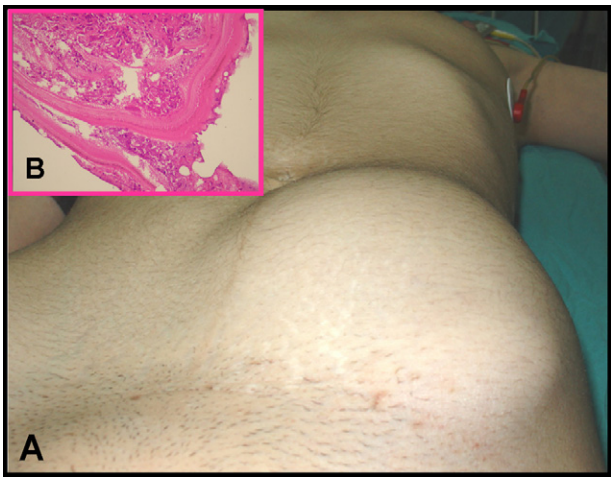

Figure 1 (A) The mass in the left lower abdomen. (B) Homogeneous cuticular membrane and inflammatory cells, HE × 100.

Case report

A 26-year-old woman was admitted to hospital with a two-month history of left lower quadrant abdominal mass, which had been detected by the patient herself. The patient had lived in a rural area where frequent contact with sheep, dogs, and other farm animals occurred. Her medical and family histories were otherwise unremarkable. She denied any change in bowel habits, diarrhea, vomiting, weight loss, chills, fever, or jaundice. Her vital signs on admission were: blood pressure 110/75 mmHg, pulse 72 bpm, and rectal temperature 37.2 °C. On clinical examination, there was a dense, slightly mobile, painless mass about 10 × 10 cm in the left lower quadrant of the abdomen. The mass was apparent by inspection (Figure 1A). Admission laboratory data were: hematocrit 36%, white blood cell count (WBC) 7.1 × 10^3/l with 1% eosinophils, total bilirubin 0.25 mg/dl, alkaline phosphatase 125 IU/l, AST 23 IU/l, ALT 28 IU/l, LDH 300 IU/l, total protein 8.3 g/dl, and erythrocyte sedimentation rate 103 mm/h. Tumor markers including CA 19-9, CA 15-3, and CEA and chest X-ray were within normal limits.

US examination revealed a mass lesion with solid and dense cystic components, measuring 9 × 10 cm, occupying the left iliac fossa. The lesion extended to the left inguinal region. A close relationship with the left ovary was noted but a thin layer of fat separated it from the lesion. A small amount of fluid was present in the rectovaginal recess. Doppler US evaluation demonstrated vascular structures inside the solid components, measuring 2—3 mm wide, with an arterial spectral pattern (Figure 2A). The lesion neighbored the left common iliac artery and vein. The external iliac artery and vein penetrated the wall of the lesion and could be followed inside the wall throughout their course. The common femoral artery and vein also neighbored the lesion.

These imaging findings suggested the lesion to be a sarcoma, and CT of the thorax and abdomen with contrast was planned to further evaluate the lesion and the presence of metastases. CT examination did not reveal any other lesions

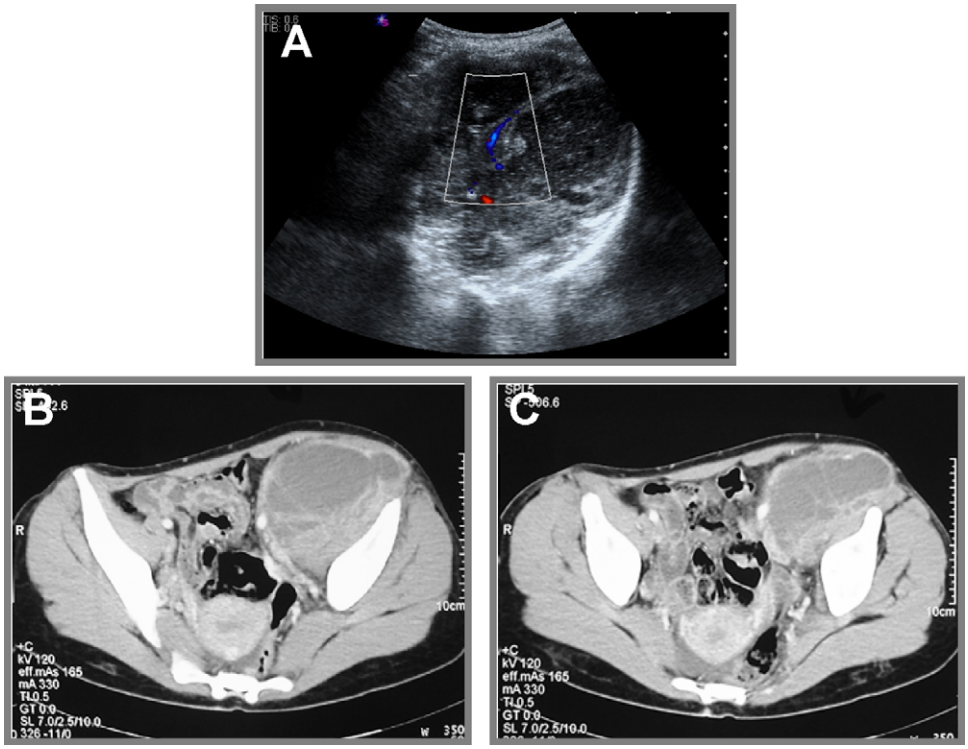

Figure 2 (A) Doppler US revealed dense cystic components and pathological vascularization at solid components. (B) CT examination demonstrates contrast enhancement at the solid components of the lesion. The left external iliac artery and vein are located at the lesion's wall. Free fluid is present at rectovaginal recess. (C) CT examination demonstrates a thin layer of fat between the lesion and the left ovary.

but demonstrated that the lesion was occupying entirely the space between the psoas muscle, the iliac wing, and the muscles of the anterior and lateral walls of the abdomen. The lesion measured 17.5 cm craniocaudally and extended from the fourth lumbar vertebra to the inguinal region at the level of the femoral neck. Cranially, the lesion neighbored the descending colon, and also fat layers around the lesion were heterogeneous. Solid components demonstrated contrast enhancement (Figure 2 B, C).

MRI demonstrated the lesion's solid components to be isointense with paravertebral muscles on T1-weighted images and hyperintense on T2-weighted images, also showing strong enhancement following the administration of contrast material (Figure 3 A—C). All of the cystic components were hypointense on T1-weighted images and hyperintense on T2-weighted images. No contrast enhancement was present at these components.

Since imaging studies suggested a retroperitoneal malignant tumor, a Tru-cut biopsy was performed. The Tru-cut biopsy revealed an eosinophilic, homogeneous cuticular membrane and inflammatory cells (Figure 1B). Since histopathologic findings indicated hydatid disease, serologic testing was performed. Positive antibody reactivity was detected in the enzyme immunoassay. A CT image of the thorax was performed for screening but was normal. The patient was prescribed albendazole before surgery.

At laparotomy, we observed that the left retroperitoneal cystic mass was bulging from the retroperitoneum and was surrounded by the sigmoid colon and mesentery of the small intestine (Figure 4A). The cyst was carefully freed from surrounding structures and the mass was punctured with a syringe; dark yellow fluid was aspirated. With the certainty of being faced with hydatid disease, a scolicidal agent was introduced and then the cyst was opened from the anterior wall. Approximately 200 ml of infected fluid was found in the cyst cavity (Figure 4B). We saw the germinative membrane and thick multisepta and removed them (Figure 4C). The left iliac artery and iliac vein were lying in the base of the adventitial cyst wall (Figure 4D). The postoperative period was uneventful. The patient was discharged ten days after the operation. At a follow-up visit 6 months after surgery, the patient remained symptom-free, and a repeat US showed no evidence of recurrence.

Discussion

HD is caused by the cystic stage of infestation by *Echinococcus granulosus*. A hydatic cyst consists of three layers: the outermost layer is adventitia, the intermediate is laminated membrane (endocyst), and the innermost is the germinal layer. Germinal epithelium is the only living part of a hydatic cyst.[1,2]

Hydatic cysts are characterized as cystic lesions with clear boundaries, which can be observed in all parts of the body.[4—6] These cystic lesions grow gradually and increase in number by means of the daughter cysts that they produce. There are no specific local or general symptoms and signs of HD, and most cases are diagnosed following incidental findings on radiographic examination for unrelated complaints. Routine blood

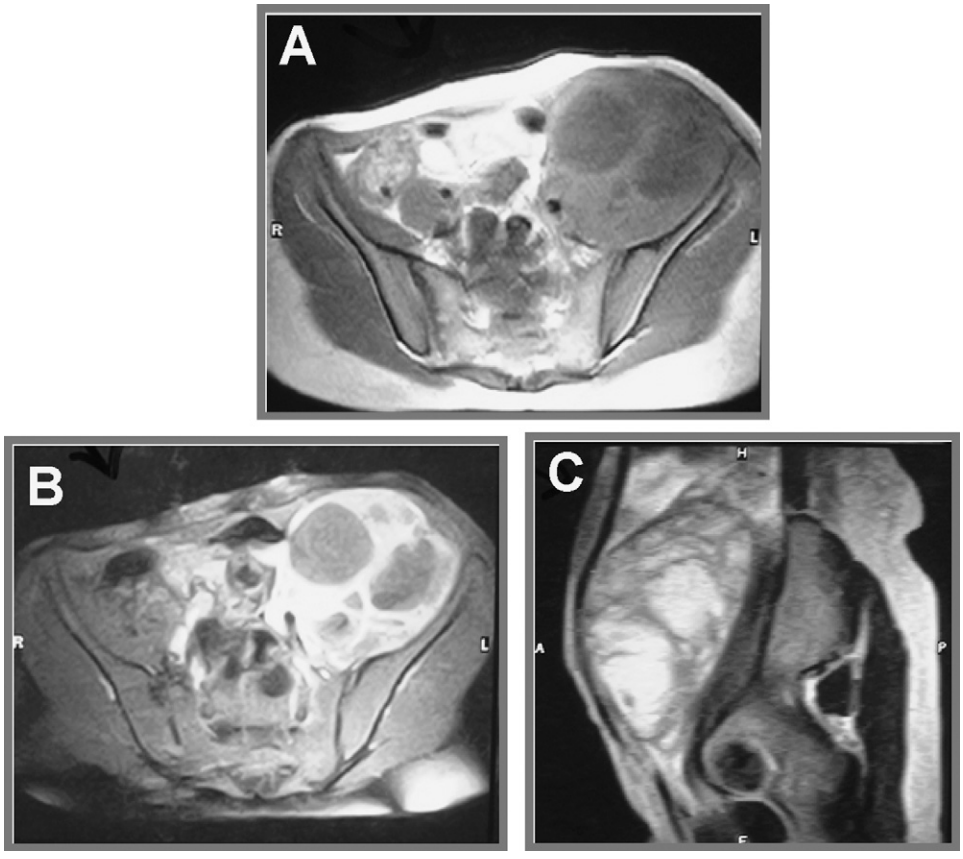

Figure 3 (A) Pre-contrast and (B) post-contrast axial T1-weighted and (C) sagittal T2-weighed MRI examinations.

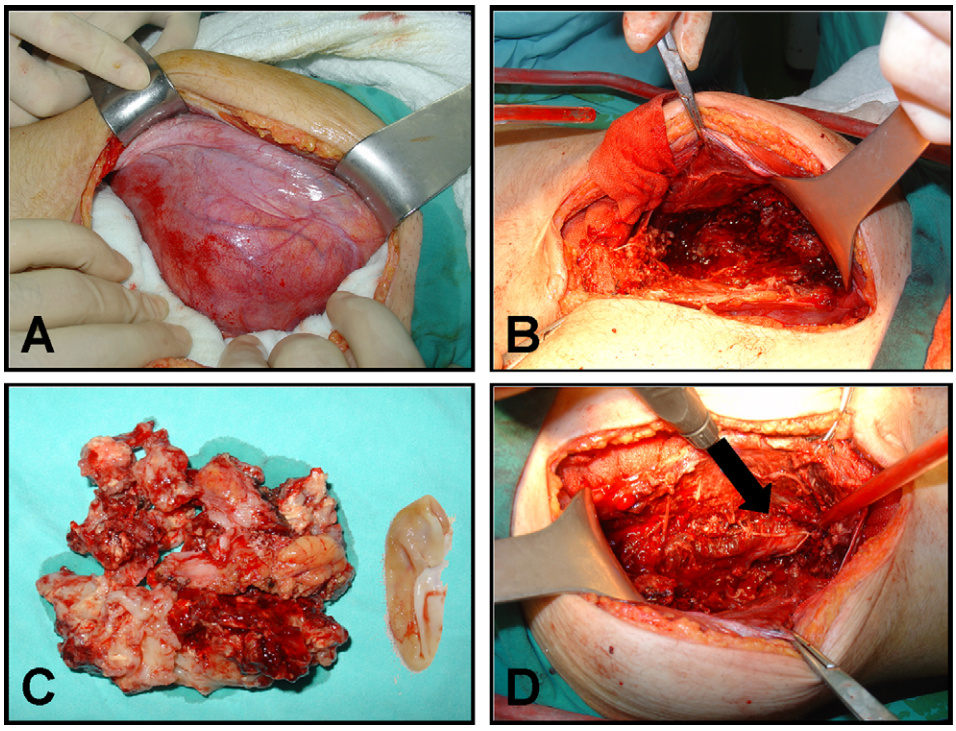

Figure 4 (A) The left retroperitoneal cystic mass. (B) The cavity of the hydatic cyst. (C) The germinative membrane and thick multisepta. (D) Left iliac artery and iliac vein in the base of adventitial cyst wall.

tests are generally normal but eosinophilia occurs in a quarter of cases. Immunodiagnostic tests may be useful for diagnosis.[1,3]

Although US and CT are sensitive in detecting hydatid cysts, HD demonstrates a variety of imaging features that vary according to growth stage, associated complications, and the affected tissue.[1,2] US evaluation of hydatid cysts may reveal a well-defined anechoic mass with or without hydatid sand and septa, a dividing septa or daughter cysts within the fluid-filled mass, calcified cysts, and undulating membranes.[1,6] CT may demonstrate a well-defined hypodense mass with enhancing septa, a cyst with round daughter cysts arranged at the periphery, calcified areas within the cyst, and also undulating membranes.[4] MRI may show the characteristic low signal intensity rim of the hepatic hydatid cyst on T2-weighted images; typically, cysts are of low intensity on T1-weighted scans and of high intensity on T2-weighted scans. The septa and cyst wall enhance after injection of contrast material, and a low signal intensity rim on T2- weighted MRI is a characteristic finding of hydatidosis. On T1- and T2-weighted MRI images, daughter cysts manifest hypointense or isointense areas compared to the maternal matrix.[1,2]

Retroperitoneal HD is usually the result of spontaneous, traumatic, or surgical rupture of a hepatic cyst. Primary retroperitoneal HD without any other organ involvement is very rare.[2–6] HD located in rare abdominal sites may lead to diagnostic difficulties, as seen in our patient, which is especially true in the case of isolated lesions in which there is no liver or lung involvement.[2,3] In conclusion HD should be considered in the differential diagnosis of all cystic masses in all anatomic locations, especially in regions of the world where the disease is endemic.

Conflict of interest: No conflict of interest to declare.

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